Case Report

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Talocalcaneal Coalition Associated with Os Sustentaculum: A Case Report

Radiology Section

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ABSTRACT

Tarsal coalition is a congenital abnormality that leads to a coalition between tarsal bones. This coalition can be bony, cartilaginous, or fibrous, with the talocalcaneal and calcaneonavicular joints commonly involved. The onset of symptoms is typically in the second decade of life and include pain, stiffness, reduced range of motion, and flatfoot deformity. Computed Tomography (CT) imaging depicts coalitions exclusively, with bony coalitions being straightforward to identify, whereas non-osseous coalitions manifest as secondary bony changes. Magnetic Resonance Imaging (MRI) shows signal changes parallel to the parent bone in cases of osseous coalition. In contrast, non-osseous coalitions exhibit variable intermediate to low signal changes, with bone marrow oedema displaying a high signal on fluid-sensitive sequences. The coalition can be located intraarticularly or extraarticularly. We present a case of an 18-year-old female who presented with right ankle pain. A CT scan of the right ankle showed an articulation between the talus and calcaneus along the medial aspect of the joint with an interspersed accessory ossicle. The extraarticular non-osseous Talocalcaneal Coalition (TCC) was diagnosed, along with the os sustentaculum located along the medial facet. In this case, we demonstrate TCC with an interspersed os sustentaculum forming an extraarticular TCC, which was detected incidentally during the evaluation of a young patient presenting with ankle pain and a visible bony deformity.

Keywords: Accessory ossicle, Ankle pain, Flatfoot deformity, Tarsal coalition

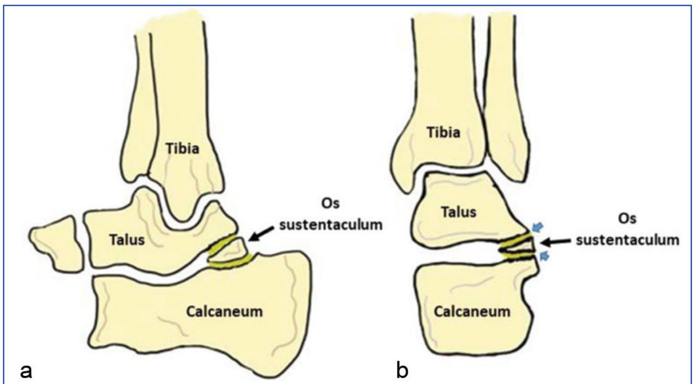
CASE REPORT

An 18-year-old female presented to the orthopaedic outpatient department with a complaint of continuous dull pain in her right ankle, which she had noticed during running over the past two weeks. She reported a history of trauma to her right great toe six months earlier, for which she received treatment at a local clinic.

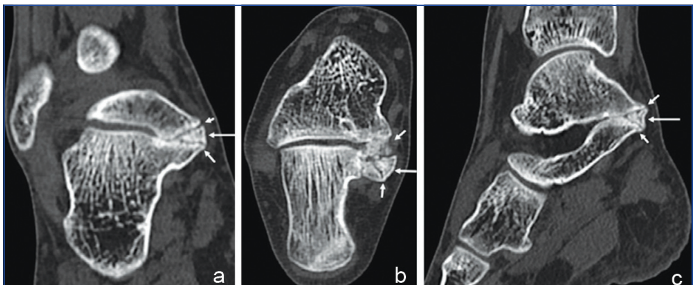
On physical examination, a bony prominence was palpated over the medial aspect of the ankle, although there was no tenderness or local rise in temperature at the site. The patient first noticed the swelling six months ago, and no signs of growth, pain, or disability in walking had been observed in the interim. No previous imaging had been performed for this issue.

The orthopaedic specialist initially ordered a radiograph of the right foot, which was inconclusive. Laboratory work was within normal limits, including complete blood count, erythrocyte sedimentation rate, and C-reactive protein. Subsequently, a Computed Tomography (CT) scan of the right ankle was requested to further examine the explanation for the clinical symptoms.

CT revealed bony remodelling of the talus's middle facet and the calcaneus adjoining the articular surface, with a triangular bony ossicle interspersed between and articulating with the calcaneus and talus [Table/Fig-1,2]. The bony abnormality was further visualised on volume-rendered Three-Dimensional (3D) images with a surface-shaded display depicting a bulge in the soft tissue [Table/Fig-3]. Bony irregularity along the inferior articular surface of the 1st metatarsal head was also noted, likely a sequela of past trauma. Based on clinical and imaging findings, a diagnosis of extraarticular non-osseous Talocalcaneal Coalition (TCC) with os sustentaculum along the medial facet was made. The patient is currently under follow-up and is planned for surgical excision.



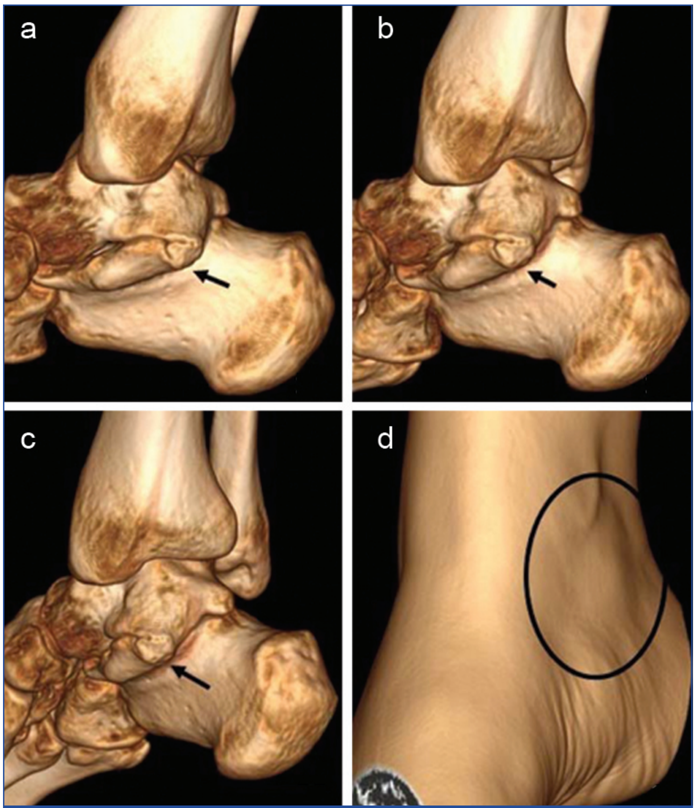
[Table/Fig-1]: Schematic diagram of the right ankle. Sagittal (a) and coronal (b) line diagrams show extraarticular Talocalcaneal Coalition (TCC) with os sustentaculum (straight arrow). There is associated bony remodelling of adjoining articular surfaces of the talus and calcaneum (blue arrowheads in 1B).



[Table/Fig-2]: Non-contrast Computed Tomography (CT) of the right ankle. Axial (a), coronal (b) and sagittal (c) images show bony remodelling along the medial aspect of the posterior facet of the talus and adjoining articular surface of calcaneum (short arrows), with a triangular Os sustentaculum interposed between the two bones (long arrows).

DISCUSSION

Tarsal coalition was first described by Zuckerland in 1877 [1]. It refers to a congenital union between two or more tarsal bones, which can be fibrous, cartilaginous, or bony [2,3]. The talocalcaneal joint is a complex hindfoot joint bearing three facets: anterior, middle, and posterior, with a tri-articular morphology [2,4]. Bony coalitions are obvious on imaging, unlike cartilaginous or fibrous variants. Associated bony abnormalities along the adjoining margins of the coalition may provide valuable diagnostic clues [2]. The incidence of tarsal coalition was considered to be around 1%. However, cadaveric studies describe incidences up to 13% and even 39% in first-degree relatives of those with known tarsal coalition, especially



[Table/Fig-3]: Volume-rendered 3D images (a-c) of the right ankle show that the triangular Os sustentaculum is visualised interposed between the talus and calcaneum along the medial aspect of the joint (arrows). The surface-shaded display image shows a soft-tissue bulge along the medial aspect of the ankle that overlies the coalition (circle).

calcaneonavicular coalition [5,6]. The underestimated prevalence can be mainly attributed to the asymptomatic nature of these lesions in early childhood [2].

On the contrary, as age progresses, the bony variants usually become noticeable due to ongoing ossification. Earlier identification of the calcaneonavicular coalition compared to the talocalcaneal type is also attributable to this ongoing ossification [2]. Tarsal coalitions are bilateral in approximately 25-50% of cases [7]. The os sustentaculum is formed by the progression of talocalcaneal fibrocartilage and its final ossification [1].

Tarsal coalitions are believed to result from a lack of mesenchymal differentiation, leading to the underdevelopment of tarsal joints. The mesenchyme develops into a cartilaginous bridge during foetal development, which later undergoes resorption over time. The persistence of this cartilaginous structure may either lead to the formation of a coalition or dedifferentiation into normal ligamentous structures [5]. Neural crest abnormalities are another implicated cause, as tarsal coalitions are frequently seen in cases with an absent fifth ray of the foot, fibular hemimelia, and spastic flatfoot in monozygotic twins [6]. Pfitzner also postulated that tarsal coalition can occur due to the incorporation of accessory ossicles within tarsal joints [6].

The onset of symptoms typically occurs in the second decade of life when there is an increase in age-related physical activity. Clinically, it can present with mid or hindfoot pain, stiffness or restricted range of movement, and spastic or rigid flatfoot [2,3]. The symptoms become more pronounced with the ossification of the coalition.

The role of imaging is to confirm the coalition, identify whether it is osseous or non-osseous, differentiate it from any anatomical variants, and assess for complications in overlying soft tissue or secondary degenerative changes in nearby joints.

Radiographs are typically the initial imaging modality used to evaluate tarsal coalitions due to their widespread accessibility, lower technical expertise requirement, and cost-effectiveness. However, they have the drawback of being a two-dimensional imaging modality with inherent limitations regarding bony overlap, which can preclude accurate diagnosis. Proper foot positioning is necessary to minimise this bony overlap, with the Harris-Beath view being a specific projection for TCC [2,5].

Radiographic findings in tarsal coalitions typically show a clear visualisation of osseous coalitions that lead to straightforward diagnoses. In non-osseous coalitions, indirect signs include narrowing of the joint space with deformed bony articular margins, which may mimic degenerative disease. Characteristic and established signs include the anteater sign (an elongated broad anterior process of the calcaneus), talar beak sign (flaring of the superior margin of the talar head), C-sign (bony continuation between the talus and sustentaculum tali) in middle and posterior coalitions, calcaneonavicular bar, laterally tapering navicular, and wide navicular. New terminologies include absent middle facet, dysmorphic sustentaculum tali, and short talar neck in TCC [2,4,7-9].

One of the first descriptions of TCC in CT and MRI was provided by Masciocchi C et al., in a four-case series [10]. Three patients were investigated using CT and MRI, which showed unilateral TCC: fibrocartilaginous in two cases and partial osseous in the third. The fourth patient, who was investigated with CT only, confirmed the presence of a bilateral fibrocartilaginous coalition.

Cross-sectional imaging modalities can accurately visualise bony and soft-tissue details using 2–3 mm reconstructed images [5]. These modalities, being three-dimensional, avoid bony or soft-tissue overlap and are not limited by the position of the foot. MRI offers the added advantage of soft-tissue evaluation.

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MRI will show bony coalitions exhibiting similar signal intensity to the adjoining parent marrow, appearing as T1 and T2 hyperintense and Short Tau Inversion Recovery (STIR) hypointense signal changes. If the non-osseous coalition is cartilaginous, it will show T1 and T2 intermediate signal changes; those with concurrent bone marrow oedema will demonstrate T2 and STIR hyperintense signal changes [2,11]. Fibrous coalitions will show low signal intensity on all sequences [10].

Various classifications have been proposed to categorise TCC into extra-articular and intra-articular types [2]. The majority of named signs are attributable to intra-articular coalitions. Posterior TCC can appear as an isolated abnormality; however, anterior coalitions are often associated with middle coalitions. Middle-facet coalitions are the most frequently encountered type of TCC [11].

Extra-articular TCC commonly occurs between the posterior margin of the sustentaculum and the posteromedial process of the talus [2]. The middle subtalar joint may be normal or hypoplastic [5]. In the cartilaginous variety of coalition, tissue is appreciable at the site of articulation, characterised by a widened osseous interval and irregular osseous articular margins. In contrast, the cartilaginous tissue will be absent in the fibrous type, and concurrent interosseous talocalcaneal ligament hypoplasia may also be seen [5]. There may be functional implications for tarsal tunnel involvement due to either bony projections or ganglion formation, potentially leading to medial plantar nerve compression. Additionally, peroneal spastic flatfoot may be encountered [5].

In severe cases, the subtalar joint may attain a ball-and-socket articular congruency [3]. Lim S et al., devised a classification system for TCC in adult patients based on CT and MRI appearances [11]. Type 1 coalitions are parallel or nearly parallel in orientation with subtalar joints, while type 2 coalitions point caudally to the subtalar joint due to overgrowth of the talus, and type 3 coalitions point cranially to the subtalar joint due to overgrowth of the calcaneum. Type 4 coalitions represent a complete bony fusion between the talus and calcaneum. Lim S et al., also highlighted the association of fractures with TCC for the first time, noting a frequency of 25%, mostly in type 1 and type 3 coalitions [11].

Rozansky A et al., developed a classification system for TCC based on CT [9]. They divided coalitions into five types, with type 1 being the most common and type 5 the least common. Types 1-3 are non-osseous, while types 4-5 are osseous, with type 4 indicating complete fusion and type 5 showing small posterior fusion. Wang A et al., also devised a similar classification system, with types 1-3 being non-osseous and type 4 representing a complete osseous coalition [12]. Each type was further divided into A, M, and P subtypes depending on the involvement of the articular facet.

Extra-articular TCC comprises one-third of all cases of Tarsal Coalition (TC), typically seen along the posteromedial aspect of the joint between the posterior sustentacular margin and the posteromedial process of the talus [13]. MRI will show corresponding abnormal masses, such as bony protrusions.

The os sustentaculum is a small accessory bone along the medial aspect of the ankle and is regarded as a normal anatomical variation. An interesting case of TCC involving the os sustentaculum was highlighted in the case series by Yun SJ et al., [1]. The os sustentaculum was considered to represent fracture fragments, as mentioned in the classification system devised by Lim S et al., and is classified as type 3-MP or type 3-P non-osseous coalition according to Wang A et al., [11,12].

Osteophytes in different locations may mimic coalition, especially along the dorsal surface of the talus and navicular, as well as at the talonavicular articular margin [7]. The appearance of these osseous excrescences at articular margins-away from typical sites of coalitions-suggestive of degenerative changes. Accessory facets along the posteromedial aspect of the subtalar joint may also resemble extra-articular TCC [13]. Other anatomical variants, such as accessory articular facets, talar ridges, medial talocalcaneal ligaments, posterior capsules of the middle subtalar joint, or capsular thickening, can be considered differential diagnoses for the extra-articular fibrous coalition, especially on MRI [2,4,7].

Treatment options vary from conservative approaches, with or without orthotic support, to surgical intervention. The initial approach to treatment involves non-operative methods, which include arch supports, casting for leg immobilisation, and positioning the foot with or without anti-inflammatory and analgesic medication [2]. However, conservative management proves effective in only about one-third of cases [12].

Surgical management is reserved for cases with minimal to unsatisfactory symptomatic improvement. Surgical options include resecting the anomalous bony articulation with or without fat interposition and excising the accessory ossicle. If excision of the bony bar is impossible or significant degenerative changes are present, arthrodesis is performed [1-3].

If left untreated, the condition will take its natural course and progress, leading to worsening pain, deformity, and limited joint mobility.

CONCLUSION(S)

Tarsal coalition is not an unfamiliar condition, with the talocalcaneal and calcaneonavicular coalitions being the most common joints involved. A sound understanding of ankle joint anatomy, particularly the hindfoot, combined with knowledge of the imaging characteristics of different coalitions across imaging modalities, is essential for accurate interpretation. A TCC can manifest with or without symptoms and should be considered a potential cause of localised ankle pain in adolescents, particularly along the medial aspect concerning the os sustentaculum. These coalitions typically manifest during late childhood or adolescence as ossification progresses, leading to restricted joint mobility and the onset of symptoms. A bony prominence along the medial aspect of the ankle can be a clinical clue to extra-articular TCC.

A correct diagnosis is essential, as oversight can lead to biomechanical dysfunction and foot disability.

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